# Successful endovascular treatment of hemosuccus pancreaticus due to splenic artery aneurysm associated with segmental arterial mediolysis

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**Hemosuccus pancreaticus, which is generally due to the rupture of a splenic artery aneursym into the pancreatic duct, is a rare cause of intermittent upper gastrointestinal hemorrhage. Segmental arterial mediolysis (SAM) is a rare arteriopathy. We report a 53-year-old man with hemosuccus pancreaticus due to a splenic artery aneurysm associated with SAM. The patient, who also had a celiac artery aneurysm affected by SAM, was successfully treated by both coil embolization and aortic stent grafting for complete coverage of the celiac artery. SAM is a very rare cause of hemosuccus pancreaticus, and endovascular treatment may be favorable for hemosuccus pancreaticus. (J Vasc Surg 2011;54:1488-91.)**

Hemosuccus pancreaticus (bleeding through the pancreatic duct into the duodenum) is a rare cause of intermittent gastrointestinal hemorrhage that cannot be easily detected by endoscopy.[1] It is usually caused by rupture of an aneurysm of a visceral artery, generally the splenic artery, associated with an inflammatory disorder of the pancreas.[2] Primary splenic artery aneurysms rarely rupture into a normal pancreatic duct. Segmental arterial mediolysis (SAM) is a rare, nonatherosclerotic, non-vasculitic arteriopathy of unknown cause characterized by development of a dissecting hematoma, aneurysm, occlusion, or hemorrhage after lysis of the arterial media.[3] We describe a case of hemosuccus pancreaticus due to a primary splenic artery aneurysm associated with SAM that was treated successfully with endovascular techniques.

## CASE REPORT

For 2 months, a 53-year-old man had recurring episodes of spontaneously resolving acute epigastric pain radiating to the back that were always followed by melena. Upper gastrointestinal endoscopy and colonoscopy yielded unremarkable results, and the source of melena had remained unknown. The patient had no history of diabetes mellitus, hypertension, ischemic heart disease, cerebrovascular disease, pancreatitis, smoking, or alcohol abuse.

At presentation, the patient was hemodynamically stable, with no abdominal symptoms and normal findings on a physical examination. The results of clinical and laboratory evaluations were as follows: body temperature, 36.3°C; heart rate, 75 beats/min; blood pressure, 135/75 mm Hg; white blood cell count, 4.1 × $10^9$/L; hematocrit, 0.40; hemoglobin, 14.4 g/dL; serum amylase, 63 U/L; and C-reactive protein, 0.7 mg/L. Coagulation panel findings were within normal limits. Except for the aneurysms, the patient had no other clinical characteristics meeting the criteria for Marfan or Ehlers–Danlos syndrome. During the entire preprocedure hospital stay, the patient's hemodynamic status was good and no blood transfusions were required.

Contrast-enhanced computed tomography (CT) showed a large 5-cm pseudoaneurysm of the splenic artery that was indistinguishable from the pancreatic body, a 2.5-cm fusiform aneurysm with a focal dissection arising from the origin of the celiac trunk, and a small fusiform aneurysm (<1.5 cm) of the superior mesenteric artery (SMA) with a region of stenosis in the proximal portion (Fig 1 and Fig 2, *A*). The renal arteries appeared normal. The findings were consistent with a diagnosis of SAM. Magnetic resonance imaging revealed that the main pancreatic duct, which was compressed by the large splenic artery pseudoaneurysm, was dilated and filled with hemorrhagic pancreatic juice (Fig 2, *B*). There was no evidence of chronic pancreatitis, so hemosuccus pancreaticus was considered the likely diagnosis. The presence of gastroduodenal collateral circulation between the SMA and the celiac artery was confirmed by CT angiography.

The patient was treated endovascularly. Through a 6F Flexor guiding sheath (Cook Medical, Bloomington, Ind) placed in the right femoral artery, a 5F Cobra catheter (Cook Medical) was inserted into the celiac artery. The splenic artery distal to the aneurysm was catheterized selectively with use of a Renegade microcatheter (Boston Scientific, Natick, Mass). The splenic artery was embolized from the distal to the proximal side of the pseudoaneurysm by using the isolation technique, with seven detachable coils employed as anchors and 27 fibered platinum coils. The celiac artery aneurysm was then embolized to the root of the splenic artery by inserting 27 detachable coils. Arteriography after coil embolization showed collateral blood flow to the liver (Fig 3, *A*), stomach, and spleen from branches originating from the SMA, with no endoleaks in either aneurysm.

To exclude inflow to the celiac artery and avoid coil compaction, a stent graft (30-mm diameter, 36-mm length; Zenith AAA endovascular graft body extension, Cook Medical) was placed in

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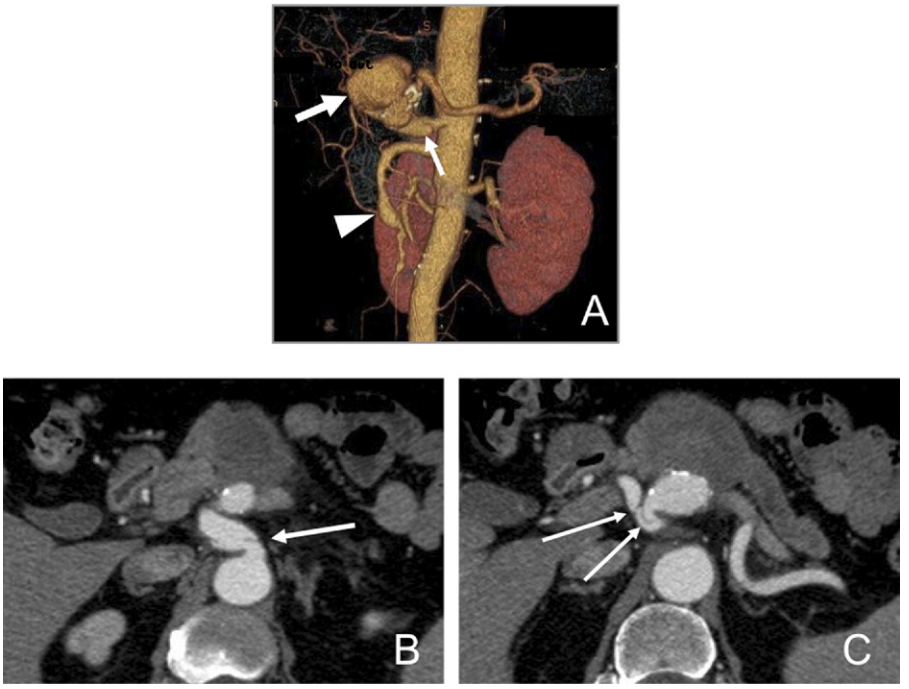


**Fig 1. A**, 3D-computed tomography (CT) angiography shows aneurysms of the splenic artery (*large arrow*), celiac artery (*small arrow*), and superior mesenteric artery (*arrowhead*), respectively. **B** and **C**, CT scan shows dissection of the celiac artery, splenic artery, and common hepatic artery at each proximal portion (*arrows*).

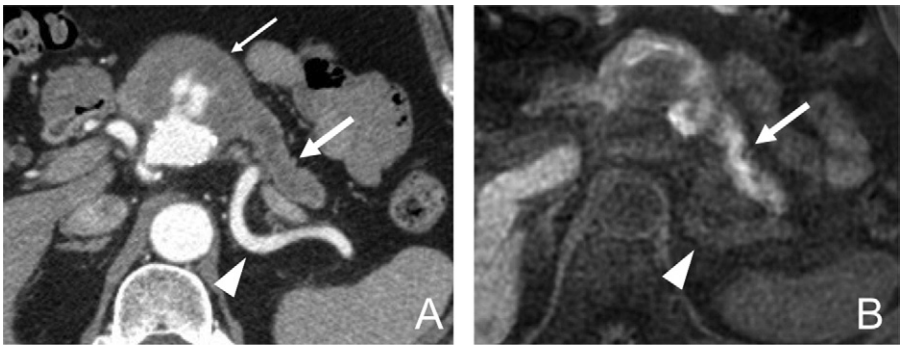


**Fig 2. A**, Axial computed tomography (CT) scan shows a large pseudoaneurysm of the splenic artery (*small arrow*) and a dilated main pancreatic duct (*large arrow*). Arrowhead shows the splenic artery. **B**, On T1-weighted fat-suppressed magnetic resonance image, a dilated main pancreatic duct was demonstrated as a hyperintense region, revealing a hemorrhage (*arrow*). Arrowhead shows the splenic artery.

the aorta just above the origin of the SMA to cover the orifice of the celiac artery completely. Because the distance between the orifice of the celiac artery and that of the SMA was short (8 mm), a balloon-expandable stent (Express LD, Boston Scientific) was placed in the SMA, positioned 3 mm into the abdominal aorta to mitigate the risk of stent graft migration and avoid closure of the SMA orifice (Fig 3, *B*) after aortic stent grafting. No blood transfusions were required during the endovascular procedure, and the estimated amount of blood loss was <200 mL.

The patient's postprocedure course was uneventful. CT angiography performed 7 days after treatment showed that the splenic artery pseudoaneurysm was thrombosed, no endoleaks had developed, and the SMA remained patent. Six months later, the patient was well, with no additional episodes of gastrointestinal hemorrhage and no evidence of pancreatitis. Repeated duplex ultrasound evaluations have revealed no signs of splenic artery recanalization.

## DISCUSSION

Gastrointestinal hemorrhage through the pancreatic duct, a rare disorder, was first described in 1931[4] and

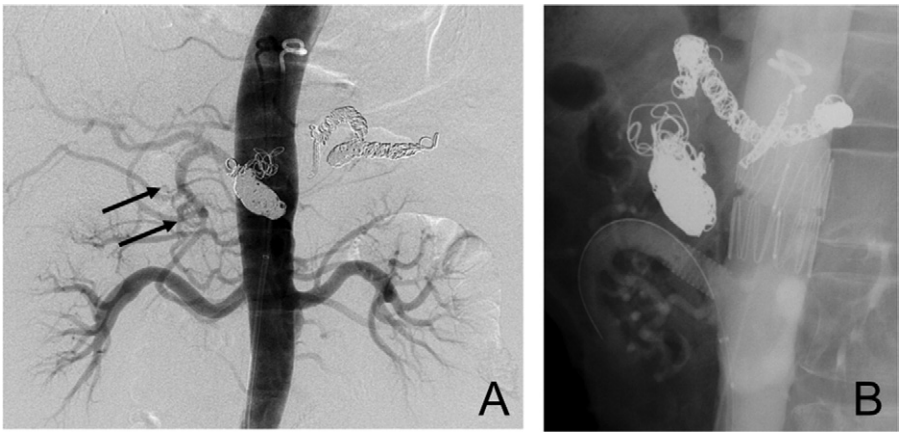


**Fig 3. A**, Arteriography after coil embolization shows complete occlusion of the splenic artery. Marked enhancement of the gastroduodenal and pancreaticoduodenal collateral network (*arrows*) supplying the hepatic artery. **B**, Lateral arteriography after aortic stent graft placement and superior mesenteric artery (SMA) stenting shows complete coverage of the celiac artery.

termed "hemosuccus pancreaticus" in 1970.[1] Pancreatic duct hemorrhage is usually due to a pancreatic pseudocyst or a peripancreatic pseudoaneurysm associated with acute relapsing or chronic pancreatitis[2]; hemorrhages with an extrapancreatic origin are rare and typically caused by a splenic artery aneurysm.[5] Hemosuccus pancreaticus can be diagnosed directly by means of endoscopic detection of bleeding from the pancreatic papilla, but this is sometimes difficult because the bleeding is often intermittent. Intermittent epigastric pain followed by melena or hematochezia within 30 to 40 minutes, as occurred in our patient, is a characteristic, and reportedly very specific,[6] manifestation of the disorder.

Splenic artery aneurysms are the most common visceral artery aneurysms. SAM is one cause of splenic artery aneurysms, although atherosclerosis is considered to be the most common.[7] SAM, first described as a distinct pathologic entity in 1976,[8] is characterized by necrosis of the outer tunica media-adventitia junction. The arterial mediolysis results in wall defects that can lead to focal dissection, aneurysm formation, stenosis, or hemorrhage caused by rupture of the artery. The etiology of SAM is poorly understood, but Slavin et al[3] postulated that the arterial lesions develop as a result of vasospasm and also suggested that SAM might be a precursor lesion of fibromuscular dysplasia. SAM usually involves more than one visceral artery, and branches of the celiac artery are frequently affected.[9] Angiographic findings are irregularity of the arterial wall, vessel widening and narrowing, occlusion or interruption, and dissection or aneurysmal enlargement.[10]

Although a definitive diagnosis of SAM requires a histopathologic evaluation of the arterial lesions, in our case, the characteristic features of SAM, including aneurysms containing regions of focal spontaneous dissection and normal-appearing renal arteries, were visualized on CT scanning. In addition, the absence of evidence of vasculitis on clinical and laboratory assessments allowed us to confirm that no vasculitic process was present and thus assume that our patient had SAM. Fibromuscular dysplasia remains a possible alternative diagnosis, but it generally occurs in the renal arteries; visceral artery involvement is uncommon. Moreover, because our patient had no findings suggestive of chronic pancreatitis or pancreatic pseudocyst, he was considered to have hemosuccus pancreaticus caused by rupture into the main pancreatic duct of a primary splenic artery aneurysm associated with SAM.

Hemosuccus pancreaticus is traditionally treated surgically, usually by distal pancreatectomy, but transcatheter selective arterial embolization has now been shown to be an effective and less invasive treatment for this disorder when it is caused by a primary aneurysm or pseudoaneurysm.[11] Naitoh et al[12] recently described a case of SAM-associated hemosuccus pancreaticus that was treated successfully with embolization. In our case, along with a large splenic artery aneurysm, the patient had a celiac artery aneurysm with dissection in the proximal portion. Therefore, to prevent type II endoleak on stent graft implantation, the celiac artery was coil embolized to the splenic artery, which had already been embolized. The stent graft was then deployed just above the orifice of the SMA and covered the celiac artery completely. An alternative to this strategy that would have avoided SMA stenting would have been to use a stent graft with a fenestration for the SMA and to land the device above the renal arteries and covering the celiac artery. In our patient, this was not possible because the proximal portion of the celiac artery was dissected; however, in patients in whom the celiac artery is a suitable landing zone, placement of an endograft from the origin of that artery into the hepatic artery after coil embolization may avoid aortic and SMA stenting. The distal SMA aneurysm in our patient was not treated because of its small size. The patient does require close long-term monitoring, however, because SAM is associ-

ated with periodic development of multiple lesions of the visceral arteries.[10]

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